SHORT REPORT

Late, Acute Presentation of a Large Brachial Artery Aneurysm Following Ligation of a Brescia-Cimino Arteriovenous Fistula

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Abstract The development of pan-arterial dilatation proximal to longstanding traumatic arteriovenous fistulae (AVF) was first described by John Hunter over two centuries ago.[1] This is thought to be due to abnormal and hyperdynamic arterial flow patterns proximal to the AVF, which renders the proximal arteries particularly susceptible to atherosclerotic change and aneurysmal dilatation.[2–4] Aneurysmal arterial dilatation has been described infrequently as a late complication of traumatic AVF formation,[5,6] however, we describe a case in which acute on chronic late brachial artery aneurysmal dilatation occurred following previous Brescia-Cimino AVF formation, which had been ligated several years earlier after renal transplantation.

Case report

A 61 year old male presented with a two week history of progressively increased swelling and aching in his left upper arm and elbow. Previously, in 1978 he had developed chronic kidney disease secondary to glomerulonephritis requiring long-term haemodialysis and had a left Brescia-Cimino arteriovenous fistula created for haemodialysis. In 1980, he underwent a successful renal transplant which precluded the need for subsequent dialysis. In 1996, the fistula was ligated as the patient was experiencing 'terrible cramps' in his hand. At operation the radial artery was repaired to maintain an adequate blood flow into the palmar arch of the hand with an end-to-end vein interposition graft. The procedure was a success and he was asymptomatic for 11 years until this episode. He presented to the unit complaining of swelling in his left arm with some discomfort. The swelling had slowly increase in size over the proceeding 2 years, but with a rapid size increase and pain over the preceding two weeks.

Examination revealed a large expansile mass in the left elbow region measuring approximately 5 cm in diameter by 10 cm longitudinally, typical of a large aneurysm. He had a prominent ulnar pulse distal to the pulsatile mass and there was evidence of previous AVF formation and ligation

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in the forearm. Allen's test revealed absent radial artery blood flow despite previous radial artery repair at the time of fistula ligation. He was normotensive and there was no evidence of distal embolisation or upper limb neurological deficit. Duplex ultrasonography revealed a 4.7 cm diameter aneurysm with intraluminal thrombus overlying the elbow arising from the brachial artery. The brachial artery was tortuous in the upper arm with localised aneurysmal dilatations measuring up to 1.5 cm diameter proximal to the large aneurysm (see Fig. 1).

A further small aneurysm at the site of the Brescia-Cimino fistula was present just above the wrist but with no flow in the radial artery proximal to this, possibly due to occlusion of the previous radial artery interposition vein graft repair. Urgent surgical repair was indicated on the basis of the rapid expansion of the aneurysm and symptoms of pain. At operation the brachial artery aneurysm was exposed through a longitudinal incision over the aneurysm and proximal and distal control achieved (see Fig. 2). The aneurysm sac was opened revealing organised and fresh thrombus and sent for histological examination. A portion of dilated cephalic vein (due to previous arteriovenous fistula) was harvested, reversed and used as an interposition graft with end-to-end anastamoses with the proximal brachial artery and the ulnar artery (see Fig. 2). The smaller segment of aneurysmal distal radial artery was also excised. Post operatively the patient made an uneventful recovery and is asymptomatic 12 months later. Histological examination demonstrated features characteristic of an aneurysm with no special features.

Discussion

Progressive arterial dilatation proximal to a ligated arteriovenous fistula with the rapid progression of aneurysm expansion is a rare occurrence.[3,5] The majority of reported cases refer to longstanding traumatic arteriovenous fistulae that were closed surgically and underwent slow progressive aneurysmal dilatation 10—15 years later. Most common cases were lower limb aneurysms — usually involving the superficial femoral and iliac arteries.[5,6]

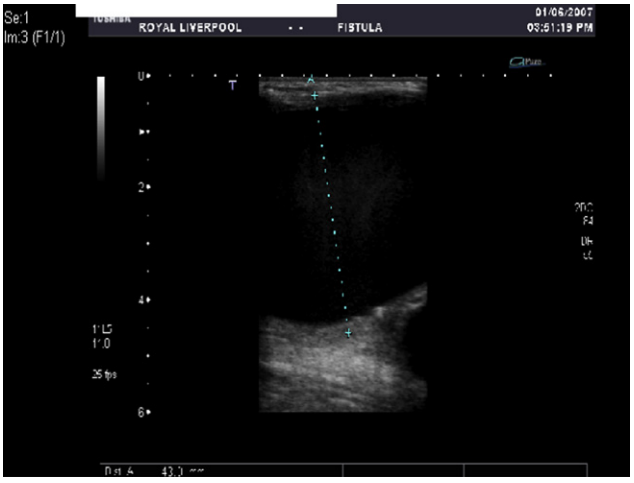

Figure 1 Duplex ultrasonography reveals 4.7 cm brachial artery aneurysm.

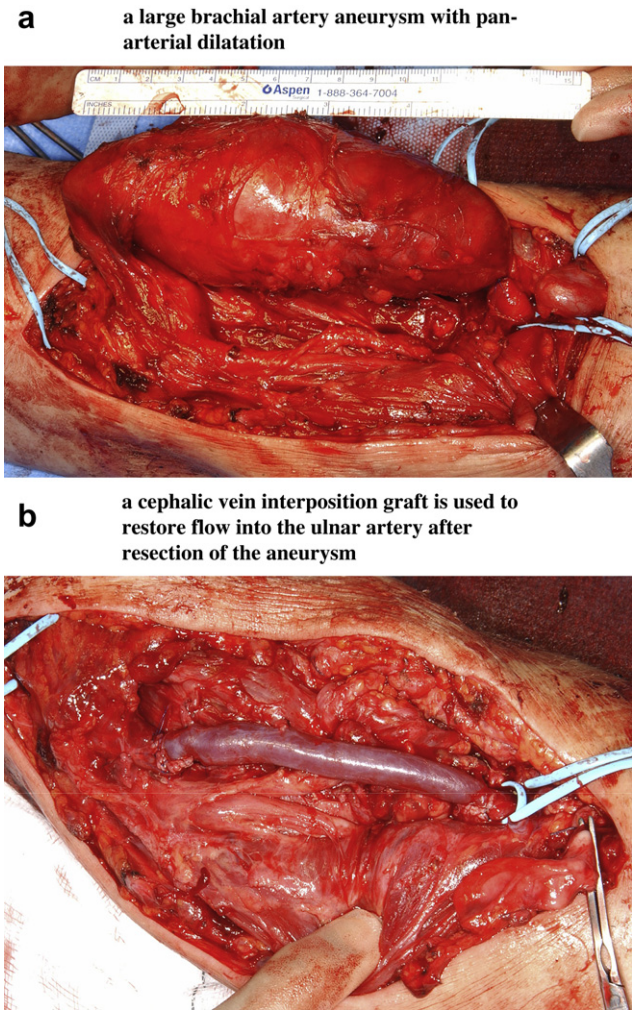

Figure 2 (a) A large brachial artery aneurysm with pan-arterial dilatation. (b) A cephalic vein interposition graft is used to restore flow into the ulnar artery after resection of the aneurysm.

The mechanisms of pan-arterial dilation proximal to the site of arteriovenous formation is postulated to be in response to increased blood flow in the vessel resulting in increased shear forces, which over a long period, causes transverse tears in the elastin fibers of the internal elastic membrane.[6,7] Without the usual healing processes, the damage starts at the fistula site and progresses proximally. The dilatation may also be mediated by an endothelium-derived humoral agent — possibly endothelium-derived relaxing factor, secreted and acting locally in response to the increase in frictional force.[5—7] Another aggravating factor may be vibrations, especially those in the audible range, which damage the elastin network and this increases the distensibility of the arterial wall.[7] In addition, it has been suggested that immunosuppression and corticosteroids could promote arterial dilatation,[2] particularly relevant here, as the patient has a long-term functioning renal transplant.

With the current drive to increase the number of transplants in the UK and Western World, it is conceivable that the number of arteriovenous fistulae being ligated in the future may rise. In this particular case, there appeared to be symptomatic reasons for fistula ligation. However, there remains uncertainty as to whether all patients who

have a successful renal transplant, should have their AVF ligated in view of the possible long-term deleterious effects of higher cardiac output than necessary, causing clinical problems in the future. Therefore, more cases of upper limb aneurysmal dilatation secondary to ligated AVF may possibly present in the future with the accompanying risks of rupture, distal embolisation and neurological deficit. We highlight the need for vigilance during long-term follow up of these patients with annual simple clinical examination, the use of duplex ultrasonography to confirm the diagnosis, and exclusion bypass surgery if clinically indicated.

Conflict of Interest/Funding

None.

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